

# *The Treatment and Postoperative Complications of Congenital Megacolon:*

## *A 25 Year Followup*

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In 1948 one of us (O.S.) proposed a new method of treatment, abdominoperineal resection, for patients with congenital megacolon. Since then, 483 patients have been treated by 13 pediatric surgeons in Chicago and Boston using this technique. Two hundred and eighty-two of the patients were last interviewed and examined more than 5 years after the resection. There were 16 postoperative deaths (3.3%) and 6 late deaths (1.2%) from enterocolitis. Both early and late complications were infrequent and are discussed in detail. Almost 90% of the patients reported that they now have normal bowel habits. None of the patients developed urinary incontinence or impotence, although ten patients (2.1%) reported permanent fecal soiling. This is the first large group of patients treated for congenital megacolon who have been followed to adulthood. The low incidence of postoperative complications and minimal frequency of long-term complications indicate that the abdominoperineal resection is a safe, effective method of treatment for congenital megacolon.

**I**N 1945 there was no cure for patients with congenital megacolon. We were fortunate to have observed three children who had colostomies performed and who were completely relieved of their symptoms. The parents requested that the colostomies be closed and treatment with drugs be tried. The colostomies were closed and the children were treated medically but, within 6 months, there was a complete recurrence of symptoms.

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The colostomies were reopened and the patients were again relieved of their abdominal distention and severe constipation. We could only conclude that the distal colon contained a physiological block, for there was no mechanical obstruction. We then studied the distal colon by multiple balloon recordings and demonstrated that in the rectosigmoid and rectum there was no peristalsis, while in the proximal colon there was normal peristalsis. Barium enema examination revealed a narrow distal colon with massive dilatation proximally.<sup>18,19</sup> The next step in this line of reasoning was that removal of the distal narrow malfunctioning colon and substituting for it the proximal dilated but physiologically intact colon should be curative.

We resected the rectum and rectosigmoid colon in 15 dogs and anastomosed the proximal colon to the rectal cuff one to two centimeters from the mucocutaneous margin. One of the dogs developed infection and was lost, but the remaining 14 animals made satisfactory recoveries and remained continent.

A similar operation was performed in 1947 on one of the three patients who had his colostomy re-established. He remained well after the colostomy closure and was continent. Since then, 482 additional patients have been resected in a similar manner. Over the past 25 years,

TABLE 1. Age at Resection and Mortality.

Age	No. Resected	(%)	Post Op Deaths	(Mortality)
0-4 Mon.	28	( 5.8)	8	(28.6)
5 Mon.-1 Yr.	128	(26.5)	3	( 2.3)
2-4 Yrs.	159	(32.9)	3	( 1.9)
5-10 Yrs.	97	(20.1)	1	( 1.0)
11-20 Yrs.	50	(10.4)	0	( 0.0)
21-50 Yrs.	21	( 4.3)	1	( 4.8)
Totals	483	(100.0)	16	(100.0)

essentially the same procedure has been used in these patients.

In a previous publication we reported a study of the symptoms, signs and diagnostic modalities used in establishing the diagnosis of congenital megacolon in 501 patients.<sup>20</sup> The present report is concerned with the postoperative complications and long-term followup on 483 patients\* who had an abdominoperineal resection as described by Swenson and Bill.<sup>17</sup> The remaining 18 patients are either awaiting resection or died before a resection could be performed.

### Method

The hospital and physicians' records of 501 patients treated for congenital megacolon at Boston Children's Hospital from 1947 to 1950, at the Tufts-New England Medical Center, Boston from 1950 to 1973 and at The Children's Memorial Hospital, Chicago, from 1960 to 1973 were reviewed. Final evaluations were obtained by traveling to various parts of the United States and Mexico and holding clinics so that these patients could be interviewed and examined. Questionnaires were not used. Information on the birth history, initial physical examination, radiologic evaluation, initial treatment including colostomy and its complications, length of aganglionic bowel, details of the resection, early and late postoperative complications and final status was obtained on each patient and was tabulated as 158 variables for the study. The data were key punched and put on a retrieval system for analysis on a Control Data 6400 Computer. The Statistical Package for the Social Sciences (SPSS) System was used to compute the frequency distribution and the Chi-square statistics for the study.

Thirteen pediatric surgeons were contributors to this series. Six of the surgeons were not trained by the senior author. In addition, 30% of the resection were performed by pediatric surgery residents.

Patients were resected in one of three ways: single-stage with no preoperative or postoperative ileostomy or colostomy; two-stage consisting of a preoperative ileos-

TABLE 2. Summary of Patient Population.

Preoperative Colostomy	202 (41.8%)
Preoperative Ileostomy	13 ( 2.7%)
Colostomy at time of Resection	7 ( 1.4%)
Ileostomy at time of Resection	17 ( 3.5%)
Colostomy after Resection	20 ( 4.1%)
Ileostomy after Resection	8 ( 1.7%)
Age at time of Resection (yrs.)	4.9±0.3*
Time taken to do Resection (hrs.)	4.3±0.1*
Postoperative Length of Stay (days)	13.8±0.8*

\*Mean and standard error.

tomy or colostomy and a takedown of the ostomy at the time of resection; and three-stage which consisted of a preoperative ostomy, resection and closure of the ostomy. Slightly more than 50% of the patients had a single-stage procedure. Less than 15% had a three-stage procedure.

The patients ages at the time of resection ranged from 8 infants less than one month of age to a 50-year-old man (Mean Age: 4.9 years) (Table 1). The mean preoperative weight was 37.2 pounds and the average time taken to do the resection was 4.3 hours. These and other patient parameters are summarized in Table 2. The proximal limit of aganglionic bowel was known in 478 of the 483 patients resected and is listed in Table 3. No patient in this study had a skip area of aganglionosis.

A final followup was obtained on 435 patients (90.1%) of those resected (Table 4). One hundred and four of the patients have been seen 16 or more years after the resection and 250 were 10 years or older when last examined.

### Results

#### Immediate Postoperative Complications (TABLE 5)

All of the patients had urinary bladder drainage postoperatively. Suprapubic drainage was used in patients under 18 months of age and urethral catheter drainage was use in older patients. Positive urine cultures were obtained on 37 (7.7%) of these patients. These were usually obtained by culturing the tip of the removed catheter and not all had clinical symptoms of urinary tract infection.

The wound infection rate of 4.6% can be attributed to

TABLE 3. Proximal Limit of Aganglionic Bowel.

	No.	(%)
Rectum	37	( 7.7)
Rectosigmoid colon	70	(14.7)
Sigmoid colon	241	(50.4)
Descending colon	67	(14.0)
Transverse colon	48	(10.0)
Ascending colon	6	( 1.3)
Terminal ileum	9	( 1.9)
Total	478*	(100.0)

\*Length of aganglionic bowel was unknown in 5 patients.

\*A previous paper by us<sup>20</sup> stated that 480 resections were performed on these 501 patients. Since publication of that paper, several key punch errors have been found to give us the corrected number of resections.

TABLE 4. *Length of Time Between Resection and Last Followup.*

Years	No.	(%)
<1	28	( 6.5)
1- 4	125	(28.7)
5-10	125	(28.7)
11-15	53	(12.2)
16-25	104	(23.9)
Total	435	(100.0)

wound contamination from a preoperative colostomy or ileostomy. Twenty of the 22 wound infections occurred in the 202 patients who had a preoperative colostomy. One of the two remaining wound infections occurred in one of the 13 patients who had a preoperative ileostomy. The corrected wound infection rate after taking down a colostomy or ileostomy at the time of resection was 9.8% (21/215). The corrected postoperative wound infection rate for the 268 who did not have a preoperative colostomy or ileostomy was 0.4% (1/268).

Six patients had dehiscence of their wound, all occurring between the fifth and eighth day after resection. Dehiscence was not related to age at resection or postoperative complications.

Leak of the coloperineal anastomosis, one of the most serious postoperative complications encountered, occurred in 24 patients (5.0%) and resulted in the death of 6 patients. The patients who had a significantly increased incidence of anastomotic leak were those with Down's Syndrome ( $P<0.001$ ). Four of the anastomotic leaks occurred in the 16 patients with Down's Syndrome. Similarly, 4 of the 37 patients with aganglionosis limited to the rectum also developed an anastomotic leak. Although this relationship is not statistically significant, the incidence of leak in patients with aganglionosis limited to the rectum was more than double that for all other lengths of aganglionic bowel.

Neither the patients age at the time of resection nor the

presence of a colostomy or ileostomy before or at the time of resection influenced the incidence of postoperative anastomotic leak (Fig. 1). We had believed that resection in the very young child carried an increased risk for this complication, but only two (7.1%) of 28 patients who were resected when they were 4 months of age or younger, developed a leak. In addition, 10 (4.7%) of the 215 patients who had a preoperative ileostomy or colostomy and two (8.3%) of the 24 patients who had an ileostomy or colostomy performed at the time of resection subsequently developed a leak. All but two of the patients who developed a leak had a diversionary ileostomy or colostomy. Two patients died before fecal diversion could be performed.

Anastomotic leak significantly increased the incidence of other serious complications. Fourteen patients had a positive blood culture after resection. Six (42.9%) of the positive blood cultures were obtained in patients who had an anastomotic leak. As expected, the incidence of pelvic and intra-abdominal abscesses was also high after leak. Eight of the 14 pelvic abscesses and 6 of the 8 intra-abdominal abscesses occurred in patients with a leak.

Thirteen patients developed a postoperative bowel obstruction requiring laparotomy within one month of the abdominoperineal resection. Over one-half of these patients developed the obstruction within one week of the resection.

Sixteen (3.3%) patients died during their hospitalization for resection. The preoperative variables, which increased the risk of postoperative death, included age at the time of resection and length of aganglionic bowel (Fig. 1). One-half of the postoperative deaths occurred in the 28 patients who were 4 months of age or younger when resected ( $P<0.0001$ ). Only one of this latter group had an anastomotic leak while 5 of the remaining 8 patients who died developed a postoperative leak. Four of the postoperative deaths occurred in the 37 patients who had aganglionosis limited to the rectum. This relationship is not statistically significant, but anastomotic leak did occur twice as frequently in patients with aganglionosis limited to the rectum than for any other length of aganglionic bowel.

#### *Late Postoperative Complications*

Late postoperative complications included rectal stricture and soiling (Table 5).

Although 30 patients (6.2%) developed a postoperative rectal stricture, only 7 required a secondary operation. The operation consisted of cutting the stricture in the two posterior quadrants. The remaining 23 patients with strictures required no treatment or rectal dilatation. Interestingly, only three of the rectal strictures occurred in patients who had an anastomotic leak. In reviewing our data, we found no predisposing factors among our vari-

TABLE 5. *Immediate and Late Postoperative Complications.*

	No.	(%)
<i>A. Immediate</i>		
1. Wound infection	22	( 4.6)
2. Wound dehiscence	6	( 1.2)
3. Anastomotic leak	24	( 5.0)
4. Sepsis	14	( 2.9)
5. Pelvic abscess	14	( 2.9)
6. Intra-abdominal abscess	8	( 1.7)
7. Bowel obstruction*	13	( 2.7)
8. Death	16	( 3.3)
<i>B. Late</i>		
1. Rectal stricture	30	( 6.2)
2. Temporary soiling	64	(13.3)
3. Death from other causes	10	( 2.1)

\*Does not include 8 obstructions occurring more than 30 days after resection.

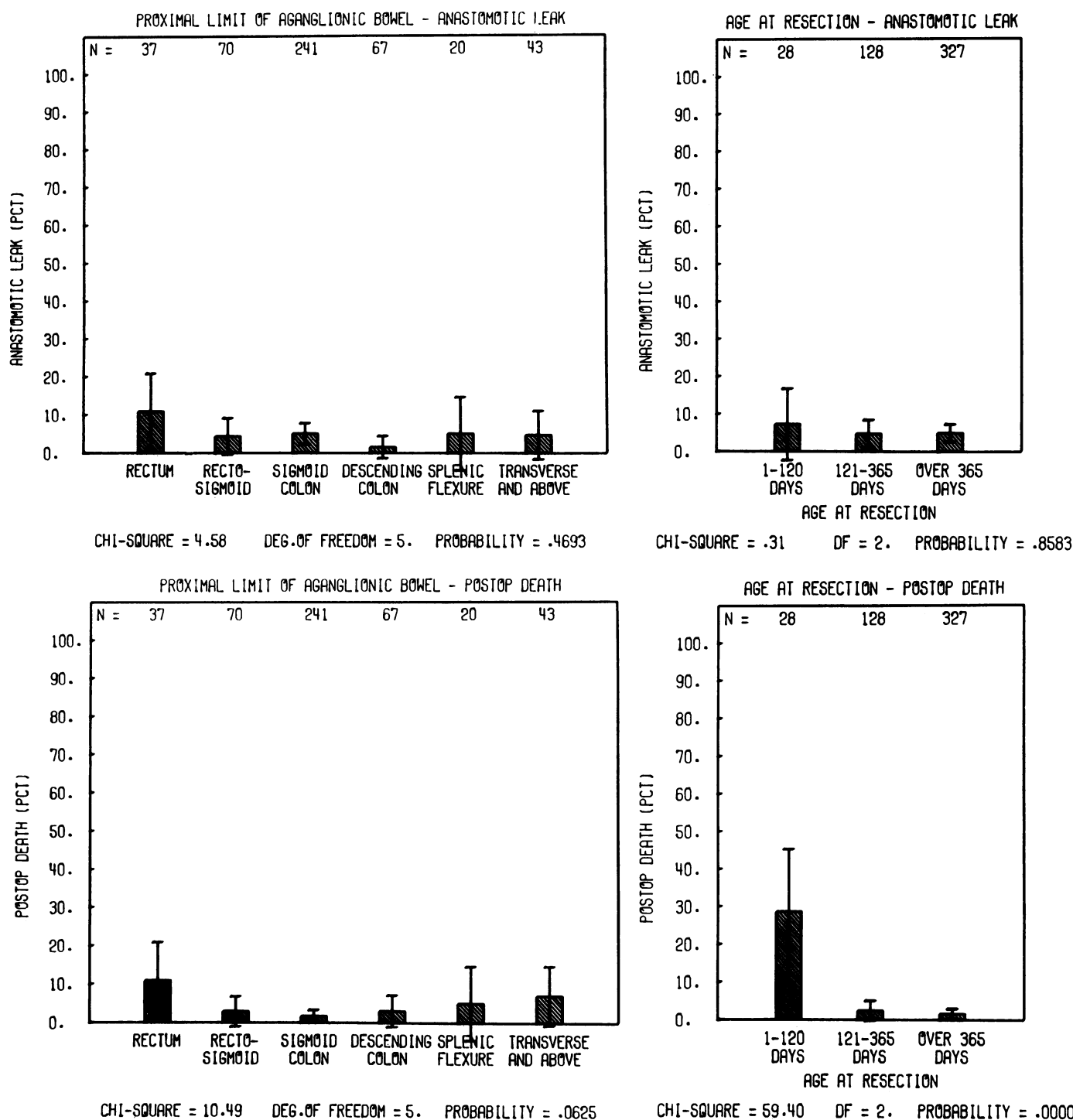


FIG. 1. Computer bar graphs comparing length of aganglionic bowel and age at resection to anastomotic leak and postoperative death. The "N" at the top of each graph represents the number of patients where information on the two variables was available. The Chi-square statistic for each relationship is given at the bottom of the graphs.

ables which increased the probability of the development of a rectal stricture.

Temporary late soiling after resection occurred in 64 patients (13.3%). Ten of these patients developed permanent soiling. The only factor which significantly in-

creased the incidence of temporary late soiling after resection was the length of aganglionic bowel. It occurred two times more frequently in patients with aganglionosis limited to the rectum than in patients with any other length of aganglionic bowel ( $P < 0.01$ ). It is of interest to

TABLE 6. *Final Patient Status.\**

	No.	(%)
1. Normal bowel habits	253	(89.7)
2. Permanent soiling	9	( 3.2)
3. Permanent colostomy or ileostomy	4	( 1.4)
4. Urinary incontinence	0	( 0.0)
5. Impotence	0	( 0.0)

\*Includes 282 patients followed for 5 or more years.

note that preoperative soiling was also noted in 12 patients with short lengths of aganglionic bowel but only two of these patients developed postoperative soiling. In 10 patients the resection corrected the preoperative soiling problem.

During the 25 years of the study, 10 patients (2.1%) died of causes unrelated to congenital megacolon or abdominoperineal resection.

### *Final Evaluation*

Although a followup final evaluation was obtained on 435 patients, only 282 patients last interviewed 5 or more years after resection will be discussed. When last seen, each patient was asked several questions about gastrointestinal and genitourinary function (Table 6).

Almost 90% stated that they have normal bowel habits. In addition, 238 patients listed the number of daily bowel movements and over 91% have one or two movements per day.

A total of 10 patients have permanent soiling. Nine (3.2%) of these have been followed for more than 5 years after resection. They range in age from 3 patients 7 years of age to one 16 year old. All were resected in the 1960's and were last seen in 1971 or 1972. No patient followed for more than 11 years has permanent soiling. Among the 9 patients with permanent soiling, one has microcephaly and mental retardation and is not trainable. An additional 3 patients have occasional staining controlled with Ducolax suppositories. Of the 5 remaining patients, one is a 15 year old who had a previous State procedure before our resection. Aganglionosis was limited to the sigmoid colon or aboral in 8 patients and extended to the cecum in the last patient.

Two patients have a permanent colostomy and two have permanent ileostomies. One of these patients, who has been followed for 22.7 years, had a total colectomy for ulcerative colitis. The second patient with a permanent ileostomy had a colectomy in 1948. He was last reviewed 23.5 years after resection when he was 31 years old. One of the patients with a permanent colostomy was 11 years old when last reviewed 6 years after resection. He developed an anastomotic leak with pelvic and intra-abdominal abscesses and required a diversionary colostomy. The colostomy has not been closed because the surgeon believes the patient has an "ulcerative colitis

type of colon with hyperperistalsis, crampy pain, mucorrhea, occult blood and anal fistulas." The second patient with a permanent colostomy was resected in 1949 and the family refused a postoperative colostomy. Eventually a colostomy was made, but he had a severe rectal stricture. He was last seen 24.5 years after resection and refuses further treatment.

Genitourinary problems were rare in the resected patients. No patient claims urinary incontinence or impotence. (The last was asked of 101 patients past puberty). The only urinary problem is in a 15-year-old boy who has permanent soiling and has nocturia if he "drinks during the evening." This patient was resected in 1961 and was last seen 11 years after resection. He required Foley catheter drainage for two months after the operation and was diagnosed as having acute pyelonephritis one month after resection.

At the final evaluation, 80 of the patients stated that they are married and cumulatively have 146 children.

### *Enterocolitis*

Enterocolitis is a poorly understood disease frequently associated with congenital megacolon and is a major cause of death in untreated patients. Almost 25% of our preoperative patients had this associated disease. The etiology of enterocolitis is unknown. Partial colon obstruction in patients with congenital megacolon has been considered a major cause of enterocolitis, but the disease also occurs in children with congenital megacolon who have normal functioning colostomies above the aganglionic bowel. When patients with acute or chronic enterocolitis are examined, the rectal sphincter is often very tight. Several years ago we proposed sphincterotomy to help these patients.<sup>16</sup> Twenty-seven were performed and our initial enthusiasm was lost because no significant benefit was obtained. Children with enterocolitis usually have a fever, abdominal distention and explosive watery green diarrhea. In the postoperative patient one may see abdominal distention and dribbling of liquid feces. X-rays of the abdomen will outline massive small and large bowel distention. The disease is common both before and after the treatment of congenital megacolon and must be diagnosed early to prevent dehydration, shock and death. Treatment is simple and effective and consists of frequent rectal irrigations with warm saline solution for two to three days. All mothers of

TABLE 7. *Incidence of Enterocolitis in Congenital Megacolon.*

	No.	(%)
Neonatal diarrhea or enterocolitis	120	(24.0)
Postoperative enterocolitis	79	(16.4)
Late enterocolitis	100	(20.7)
Late death from enterocolitis	6	( 1.2)

patients with congenital megacolon are warned about this problem and instructed in the technique of rectal irrigations.

Diarrhea or enterocolitis after birth occurred in 120 patients (Table 7). These patients had a significantly increased incidence of late enterocolitis ( $P < 0.01$ ) occurring more than one or two months after resection, but not enterocolitis occurring in the immediate postoperative period. Over 28% of the patients with neonatal diarrhea or enterocolitis developed late enterocolitis, but only 16.2% of those patients without a history of neonatal diarrhea or enterocolitis eventually developed late enterocolitis.

Seventy-nine patients developed immediate postoperative enterocolitis. A significant per cent of these patients (38.0%) eventually developed late enterocolitis ( $P = 0.0001$ ). Only 17.8% of the patients without immediate postoperative enterocolitis developed late enterocolitis.

One hundred patients (20.7%) developed late enterocolitis. Sixty-eight of these patients (14.1%) required hospitalization for treatment of the enterocolitis. The incidence of late enterocolitis requiring hospitalization was most common in the first year after resection when 51 (75.0%) patients had their last episode. Of the remaining 17 patients, 12 (17.6%) had their last attack during the second year after resection. No patient had late enterocolitis requiring hospitalization more than 5.4 years after resection.

The age of the patients at the time of resection also influenced the incidence of immediate postoperative and late enterocolitis. Enterocolitis occurred in 30% to 40% of the children who were resected when they were 4 months of age or younger and in 10% to 20% of the older children.

Six patients (1.2%) died from enterocolitis during the period three to 36 months after resection. All of these patients had had immediate postoperative and/or late enterocolitis. Only one had had diarrhea or enterocolitis as a newborn.

### Discussion

This long-term study demonstrates the safety and efficacy of abdominoperineal resection for the definitive treatment of congenital megacolon. It is difficult to compare our results with other reported series using the Duhamel or Soave operations because of lack of comparable data.

Our postoperative mortality of 3.3% includes 8 of the 28 patients who were 4 months or younger when they were resected. We no longer perform a resection on patients less than 6 months of age or on patients who weigh less than 20 pounds. If these 28 patients are excluded when calculating mortality, we obtain a corrected death rate of 1.8% (8/455).

Enterocolitis occurred frequently in our pre and post-operative patients and requires further study. A more detailed review of these patients will be performed in the near future to give us more insight into this complex and difficult problem.

Anastomotic leak, which was a major cause of death in patients who were over 4 months of age when resected, was also associated with a marked increase in morbidity. This study suggests that patients with a short length of aganglionic bowel or with Down's Syndrome carry an increased risk for this complication. Other important factors predisposing patients to an anastomotic leak, such as an inadequate blood supply at the resected margin or tension at the suture line, could not be quantitated for computer review.

Our data are in agreement with Fazio<sup>8</sup> and Burman,<sup>5</sup> who believe that a protective ostomy does not influence the incidence of leaks in distal colon anastomoses. While we do not believe that a protective ostomy will prevent leaks, it may be lifesaving in preventing continued contamination of the pelvis and the peritoneal cavity after a leak is discovered.

Aylett,<sup>1</sup> in a series of 300 low colonic anastomosis in patients with ulcerative colitis, reported a 12.4% incidence of leak. Burman<sup>5</sup> reported a 32% incidence of leak in low colonic anastomosis for Crohn's disease. Our rate of 5% compares favorably with these. However, our patients are not comparable particularly with the Crohn's disease patients, since these latter patients have a problem in healing. In 163 ileorectal anastomoses for a variety of conditions at the Cleveland Clinic, the leak rate was 1.2%. Polacek<sup>14</sup> reported a leak rate of 3% in 270 colon anastomosis for carcinoma. Our anastomosis is below the peritoneal floor and one would expect a higher incidence of leaks. It would seem that a 5% leak rate is reasonable.

Rectal stricture is often mentioned as a major complication of this type of operation, but severe rectal stricture occurred in only one of our patients. The mother of this patient refused to permit a prompt postoperative colostomy when a leak at the anastomosis occurred. In all the subsequent leaks, a prompt colostomy was made in all patients when the leak was detected. None of these patients developed a severe stricture which required a second resection. In a group of patients with strictures sent to us for a second resection, all had colostomies delayed weeks or months after the leak occurred.<sup>4</sup> We firmly believe an immediate colostomy is mandatory after a leak has been identified.

Duhamel<sup>7</sup> described a side to side anastomosis of normal colon to the aganglionic rectum using a spur-crushing clamp. This operation has been modified by Martin. His first report in 1962 described a modification of the Duhamel operation and he expressed satisfaction with

the results in seven patients.<sup>12</sup> He has subsequently modified this to a double anastomosis to eliminate the troublesome blind rectal pouch.<sup>13</sup> Others have modified this by using a stapling device to achieve the side to side anastomosis.

Soave<sup>15</sup> advocates an endorectal operation, a technique first described by Hockenegg in 1900.<sup>9</sup> The rectal mucosa is removed down to the mucocutaneous margin and the normal colon pulled through this and left protruding for ten to 14 days. Boley<sup>3</sup> had difficulty with this procedure and proposed making an anastomosis at the mucocutaneous margin. He reports 8 cases with satisfactory results. Presumably, because of some problems with the retained aganglionic rectal cuff, Marks<sup>11</sup> has modified the operation by cutting the muscular cuff posteriorly. Deodhar<sup>6</sup> has reported his experience after doing 20 Soave operations, including 8 with the Boley modification. He had a 15% mortality rate and complications in 65% of the patients. Klotz<sup>10</sup> reported his complications after doing three Soave and 16 Boley operations with anal strictures in 4, perineal excoriation in 5 and mucosal prolapse in 3. These are small groups of patients and consequently not suitable for a meaningful comparison with our results.

Resection of the aganglionic colon with a pull-through anastomosis is a difficult operation. Yet, if a well trained surgeon has an opportunity to observe the technical details of the operation and then perseveres, good results can be obtained. Our results and those of Weitzman,<sup>21</sup> who reported 41 cases with a low complication rate and good results, are evidence for this. Bennet<sup>2</sup> has a large series of resections with pull-through anastomosis with good results.

The final status of 282 of our patients followed for 5 or more years demonstrates the excellent long-term results of abdominoperineal resection with a pull-through anastomosis. Almost 90% of the patients followed for five to 25 years have normal bowel habits and none have urinary incontinence or impotence. Of the 29 patients without normal bowel habits, only 13 have the serious problems of permanent soiling or fecal diversion. The remaining 16 patients complained of recurrent diarrhea or constipation requiring enemas or laxatives.

The one helpful bit of information that has improved the treatment of congenital megacolon, was that the aganglionic distal colon was a physiological block. We still believe that the most enduring good results are obtained when this is removed and not partially left in place.

## Acknowledgments

We wish to thank Nan MacMillan and Ruth Kobbe for their valuable assistance in programming a major portion of this study.

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## DISCUSSION

DR. CLIFFORD D. BENSON (Detroit): It is a distinct pleasure for me to discuss the paper of Drs. Swenson, Fisner, Sherman and Cohen. Dr. Swenson has had the unique experience of originating an operative

procedure based on sound experimental work and applying it to patients with Hirschsprung's Disease with curative results which have stood the test of time throughout the surgical world.

Our interest in Hirschsprung's Disease dates from 1948 when Dr. Solzer and Dr. Wilson at the Children's Hospital in Michigan reported